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*Published in:*  
American journal of speech-language pathology

*DOI:*  
[10.1044/2018\\_AJSLP-17-0013](https://doi.org/10.1044/2018_AJSLP-17-0013)

**IMPORTANT NOTE:** You are advised to consult the publisher's version (publisher's PDF) if you wish to cite from it. Please check the document version below.

*Document Version*  
Publisher's PDF, also known as Version of record

*Publication date:*  
2018

[Link to publication in University of Groningen/UMCG research database](#)

*Citation for published version (APA):*  
Terband, H., Spruit, M., & Maassen, B. (2018). Speech Impairment in Boys With Fetal Alcohol Spectrum Disorders. American journal of speech-language pathology, 27(4), 1405-1425.  
[https://doi.org/10.1044/2018\\_AJSLP-17-0013](https://doi.org/10.1044/2018_AJSLP-17-0013)

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## Research Article

# Speech Impairment in Boys With Fetal Alcohol Spectrum Disorders

Hayo Terband,<sup>a</sup> Manon Spruit,<sup>a,b</sup> and Ben Maassen<sup>c</sup>

**Background:** Fetal alcohol spectrum disorders (FASD) are a highly prevalent spectrum of patterns of congenital defects resulting from prenatal exposure to alcohol. Approximately 90% of the cases involve speech impairment. Yet, to date, no detailed symptom profiles nor dedicated treatment plans are available for this population.

**Purpose:** This study set out to chart the speech and speech motor characteristics in boys with FASD to profile the concomitant speech impairment and identify possible underlying mechanisms.

**Method:** Ten boys with FASD (4.5–10.3 years old) and 26 typically developing children (4.1–8.7 years old; 14 boys, 12 girls) participated in the study. Speech production and perception, and oral motor data were collected by standardized tests.

**Results:** The boys with FASD showed reduced scores on all tasks as well as a deviant pattern of correlations between production and perception tasks and intelligibility compared with the typically developing children. Speech motor profiles showed specific problems with nonword repetition and tongue control.

**Conclusions:** Findings indicate that the speech impairment in boys with FASD results from a combination of deficits in multiple subsystems and should be approached as a disorder rather than a developmental delay. The results suggest that reduced speech motor planning/programming, auditory discrimination, and oral motor abilities should be considered in long-term, individually tailored treatment.

Prenatal exposure to alcohol can have a teratogenic effect on the developing embryo and fetus, including growth deficiencies, physical malformations, and central nervous system (CNS) anomalies (e.g., Jones, 2011; Jones & Smith, 1973; Kodituwakku, 2007; O'Leary, 2004). The resulting patterns of congenital abnormalities are termed *fetal alcohol spectrum disorders* (FASD; Bertrand, Floyd, & Weber, 2005; Sokol, Delaney-Black, & Nordstrom, 2003), ranging from confined alcohol-related neurodevelopmental disorders or birth defects to fetal alcohol syndrome (FAS; Jones, Smith, Ulleland, & Streissguth, 1973) as its most severe form (e.g., Hoyme et al., 2005; May et al., 2014). Approximately 90% of the children with FASD display a speech impairment that has been described in general terms as problems involving fluency, articulation, nasality, and

word formulation (Becker, Warr-Leeper, & Leeper, 1990; Church, Eldis, Blakley, & Bawle, 1997; Manning & Hoyme, 2007). However, the speech characteristics have not yet been described in detail, the impairment is not well known among speech-language pathologists (SLPs), and the disorder is often not recognized (Chasnoff, Wells, & King, 2015; Williams & Smith, 2015). This study set out to examine the phonological and speech motor symptoms in children with FASD to characterize the concomitant speech impairment in FASD. This information is paramount for the development of effective treatment programs and might enable early detection and intervention.

## Prevalence

Reported birth prevalence estimates of FASD vary widely depending on cultural and demographic aspects (Roozen et al., 2016). General numbers range from between 9 and 10 per 1,000 in most samples (Manning & Hoyme, 2007), 15 per 1,000 in families with foster children (Astley, Stachowiak, Clarren, & Clausen, 2002), and up to 39–46 per 1,000 in specific communities (May et al., 2000; O'Leary, 2004). However, recent studies suggest these estimates might be too conservative, yielding numbers that are considerably higher. May and colleagues (2014, 2015) reported prevalence

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Editor-in-Chief: Julie Barkmeier-Kraemer

Editor: Carol Miller

Received January 26, 2017

Revision received August 1, 2017

Accepted May 11, 2018

[https://doi.org/10.1044/2018\\_AJSLP-17-0013](https://doi.org/10.1044/2018_AJSLP-17-0013)

**Disclosure:** The authors have declared that no competing interests existed at the time of publication.

estimates of 11–25 per 1,000 (May et al., 2015) and 24–28 per 1,000 (May et al., 2014) in representative U.S. communities (see also Fox et al., 2015). In remote rural areas in South Africa and Australia, numbers reach as high as 64 per 1,000 (Urban et al., 2015), 120 per 1,000 (Fitzpatrick et al., 2015), up to 135–208 per 1,000 (May et al., 2013). Among the specific group of foster children and orphans, prevalence estimates can be even higher, ranging around the world from 40 per 1,000 up to 521 per 1,000 for children from Eastern Europe (Lange, Shield, Rehm, & Popova, 2013). On the basis of similar samples regarding population and geographical area, prevalence of FASD is high compared with other congenital syndromes, even when assuming a conservative estimate (see Figure 1).

### Clinical Characteristics of FASD

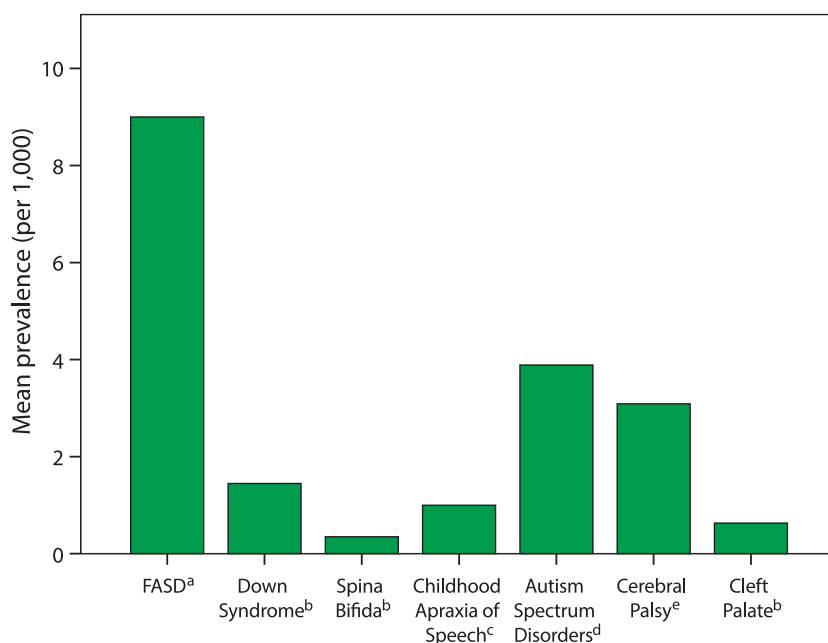
The more severe forms of FASD involve anatomical abnormalities. The physical malformations include growth deficiency and craniofacial dysmorphology. Cardinal facial features are small palpebral fissures (opening between the eyelids), a smooth philtrum, and a thin vermilion border of the upper lip lacking tubercle or Cupid's bow (e.g., Jones, 2011; Jones & Smith, 1973; Kodituwakku, 2007; O'Leary, 2004). Further common orofacial features are malocclusion of teeth; a heightened palate; midfacial, maxillary, and mandibular hypoplasia (undersized cheekbones, eye sockets, maxillary bones, or jaw); a flattened, short, or low nose bridge; epicanthal folds (a skin fold of the upper eyelid covering the inner corner of the eye); and ear anomalies (lower

positioned and deviant-shaped auricle, so-called “railroad track ear”; Abell et al., 2016; Jones et al., 2010; Sampson et al., 1997; Suttie et al., 2013).

Neuroanatomical abnormalities include microcephaly (small head circumference due to brain underdevelopment) as well as structural anomalies across the entire CNS. Such structural CNS abnormalities may comprise hypoplasia of cortical (low gray-matter volume), subcortical (including underdevelopment of cerebellum and basal ganglia, especially the caudate nuclei), and white matter (partial or complete absence of the corpus callosum) structures (Archibald et al., 2001; Chen, Coles, Lynch, & Hu, 2012; Mattson et al., 1996; Norman, Crocker, Mattson, & Riley, 2009; Roebuck, Mattson, & Riley, 1998).

In terms of cognitive functioning, an FASD has been associated with deficits in attention, learning and executive functions, mental retardation, fine and gross motor difficulties, hearing disorders, and language and speech impairments (Abkarian, 1992; Becker et al., 1990; Church et al., 1997; Cone-Wesson, 2005; Lewis et al., 2015; O'Leary, 2004). There is a high overlap with other neurodevelopmental disorders—in particular, attention-deficit/hyperactivity disorder may involve similar deficits in inhibition and information processing (Landgren, Svensson, Strömberg, & Grönlund, 2010; O'Malley & Nanson, 2002), but also links with autism spectrum disorders have been established (O'Malley & Rich, 2013)—and in practice, an FASD often remains unrecognized or is misdiagnosed (Chasnoff et al., 2015).

**Figure 1.** Prevalence of fetal alcohol spectrum disorder (FASD) in comparison with other congenital syndromes based on similar samples regarding population and geographical area: <sup>a</sup>Manning and Hoyme (2007), <sup>b</sup>Parker et al. (2010), <sup>c</sup>American Speech-Language-Hearing Association (2007), <sup>d</sup>Baird et al. (2006), and <sup>e</sup>Arneson et al. (2009).



## Speech Impairment in FASD

To date, the specific characteristics of the concomitant speech impairment in FASD have not been described in detail. The speech impairment in FASD is suggested to be the result of a combination of CNS, hearing, and oral motor (including craniofacial abnormalities) defects and is generally described as “misarticulations persisting longer than what is appropriate for their chronologic age” (Church & Abel, 1998, p. 89). In other words, the speech problems are often assumed to reflect a developmental delay. The clinical impression of SLPs who have experience with this disorder, however, is that a developmental delay does not cover the whole story (see also Becker et al., 1990; Manning & Hoyme, 2007). From a theoretical viewpoint, the combination of neurocognitive deficits—including oral motor and hearing deficits—raises suspicion that speech development might be not only delayed but also deviant in children with FASD.

### Aim of This Study

This study comprised a detailed investigation of speech and speech motor characteristics in boys with FASD based on an array of standardized speech production and perception, and oral motor assessments. The focus of the study on boys with FASD came out of necessity. We did not select on gender when recruiting participants and approached the parents/caretakers of both boys and girls with FASD. However, the cases in which parents/caretakers and children were willing to participate included only boys. Whether this reflects gender-related differences in the prevalence of FASD or in the expression of adverse effects of prenatal alcohol exposure is an interesting question worthy of further investigation but is beyond the scope of this study.

Our goal was to create a detailed profile of the concomitant speech impairment in children with FASD by investigating commonalities and individual differences in phonological and speech motor development as compared with typically developing (TD) children. First, we set out to create a detailed profile of the symptomatology through a quantitative and qualitative analysis of speech errors using standardized speech tasks. We then aimed to establish whether speech development in boys with FASD is delayed or (also) deviant by analyzing how the speech profile of the boys with FASD differs from the profile observed in typical development. If their development was found to be (also) deviant, this would mean in clinical terms that their speech impairment should be approached therapeutically as a disorder rather than as (just) a developmental delay. There are two reasons why we used a comparison group of TD children in this study. First, no reference norms are available for most of the assessments and tests that we used in this study. Second, an important part of the speech profiling that we pursued involves the investigation of patterns of correlations between multiple outcome measures. Finally, we sought to identify possible underlying mechanisms of the concomitant speech impairment in FASD as to inform

the choice and possible future development of treatment programs and methods for early detection and intervention.

First, we investigated speech intelligibility as an indicator of the severity of the impairment experienced in daily life. Next, a quantitative segmental analysis of word production accuracy on standardized speech tasks was made, resulting in inventories of phonetic accuracy and phonological error characteristics. To establish whether development is delayed or deviant, we further investigated these inventories in detail on measures of phonological complexity and phonological processes. These analyses reflect two dimensions of speech development: the order in which speech sounds are typically acquired and the way speech sounds are typically produced during the process of acquisition. Finally, the identification of possible underlying mechanisms was based on task comparisons and on correlations between speech production, and auditory discrimination and oral motor functional performance. Hearing disorders associated with FAS comprise four types: delays in auditory maturation, sensorineural hearing loss, intermittent conductive hearing loss due to recurrent serious otitis media, and central hearing loss (e.g., Church & Abel, 1998). Hearing deficits are known to be common in FASD, although the numbers on prevalence reported in the literature vary widely. Cross-sectional studies report that 21%–77% of the children with FAS suffer a form of hearing loss (e.g., Church & Abel, 1998; Kvigne et al., 2004; Rössig, Wässer, & Oppermann, 1994). In addition to hearing status, in this study, we also investigated the potential role of reduced auditory feedback on a functional level by measuring auditory discrimination and evaluating a possible relation with speech symptoms. Similarly, oral motor abilities were measured to investigate the potential role of craniofacial abnormalities.

## Method and Materials

### Participants

Ten boys aged 4.5–10.3 years ( $M = 7.4$  years,  $SD = 1.9$  years) with FASD and 26 TD children aged 4.1–8.7 years ( $M = 5.6$  years,  $SD = 1.4$  years) participated in the study. Written consent was obtained from all parents or caretakers before starting the study. The TD children were recruited through local schools and acquaintances as part of a larger study (Nijssen, van Brenk, & Terband, 2015; Terband & van Brenk, 2015; Terband, van Brenk, & van Doornik-van der Zee, 2014; van Doornik, Gerrits, McLeod, & Terband, 2018). The boys with FASD were recruited through speech pathologists and the Dutch FASD foundation. FASD diagnoses were made by a specialized pediatrician following the criteria defined by Manning and Hoyme (2007). Background data are presented in Table 1, and a description of craniofacial characteristics is presented in Table 2. Information about hearing status was available for eight of the boys with FASD. Three had a history of hearing problems and had mild hearing loss (pure-tone threshold between 25 and 40 dB at least one frequency), whereas the remaining five did not have a history of hearing

**Table 1.** Overview of the children with fetal alcohol spectrum disorder (FASD) who participated in the study.

Child ID	Diagnosis	Age (years)	Gender	Family situation	Hearing status	Speech therapy
FASD1	pFAS	9.9	M	Biological mother	No information available	Yes
FASD2	FAS	5.5	M	Adopted	Mild hearing problems reported at a young age; borderline mild hearing loss, right ear only (hearing threshold of 30 dB HL at 4000 Hz)	No
FASD3	FAS	6.8	M	Adopted	History of multiple otitis media; mild hearing loss binaurally	Yes
FASD4	FAS	6.5	M	Adopted	No history of hearing problems; no recorded hearing loss	No
FASD5	FAS	4.5	M	Foster parents	No history of hearing problems; no recorded hearing loss	Yes
FASD6	FAS	10.3	M	Adopted	No history of hearing problems; no recorded hearing loss	No
FASD7	FAS	6.7	M	Adopted	No history of hearing problems; no recorded hearing loss	No
FASD8	FAS	7.2	M	Adopted	History of multiple otitis media; mild hearing loss binaurally	Yes
FASD9	FAS	5.7	M	Foster parents	No history of hearing problems; no recorded hearing loss	No
FASD10	FAS	8.8	M	Biological mother	No information available	Yes

Note. pFAS = partial fetal alcohol syndrome; M = male; FAS = fetal alcohol syndrome.<sup>a</sup>

<sup>a</sup>pFAS is a diagnostic classification for patients with confirmed prenatal alcohol exposure who present with central nervous system damage (structural, neurological, and/or functional impairment) and some but not all of the physiological symptoms of full-blown FAS (e.g., Hoyme et al., 2005; May et al., 2014).

problems and did not have an indication of hearing loss recorded during the regular governmental hearing screening at the age of 4–5 years. Half of the boys with FASD had a history of or still received speech therapy. The TD group comprised 14 boys and 12 girls. All children in the TD group had normal hearing (pure-tone thresholds not exceeding 25 dB HL) and normal speech-language development and intelligence (scores not less than 1 *SD* below population average). Although earlier studies with identical or similar outcome measures as used in this study have not found or reported any between-gender differences (e.g., Beers, 1995; Rvachew & Grawburg, 2006; Smith, Goffman, & Stark, 1995; Terband, Maassen, Van Lieshout, & Nijland, 2011), a possible effect of gender on any of the outcome measures in the TD group was explored in a series of statistical tests. As results did not reveal any main or interaction effect of gender (or a trend thereof) for any of the outcome measures,

we concluded that gender differences in the TD group could be safely ignored for the remaining analyses. Furthermore, groups were not equivalent in mean age,  $t(34) = -2.844$ ,  $p = .007$ . Because the participants in the group with FASD were older compared with the participants in the TD group, the bias of higher chronological age can be accepted safely as it does not inflate the risk of a Type I error (incorrectly rejecting the absence of group differences). In addition, age was entered as a covariate in the remaining analyses.

### Data Collection

Speech production and perception, and oral motor data were collected by standardized tests. Intelligibility was assessed using the Intelligibility in Context Scale (ICS-NL; McLeod, Harrison, & McCormack, 2013). Speech production was assessed by the Computer Articulation Instrument (CAI;

**Table 2.** Description of craniofacial and orofacial anatomical characteristics of the boys with fetal alcohol spectrum disorder (FASD) who participated in the study.

Child ID	LPhG <sup>a</sup>		Dental information	Nose bridge	Other information
	Lip/Philtrum	Palatum			
FASD1	3/3	Flat and high	Secondary teeth, light malocclusion (Class 2)	Typical	—
FASD2	5/5	High	Primary teeth, malocclusion (Class 2)	Short	—
FASD3	4/4	High	Crossbite, secondary incisors and canine, primary molar	Short	Short lingual frenulum
FASD4	3/3	High	Primary teeth	Typical	—
FASD5	5/5	High	Primary teeth	Short	No tongue lifting possible
FASD6	5/5	Typical	Primary molars, secondary incisors and canines	Typical	—
FASD7	5/5	High	Primary molars, secondary incisors and canines	Typical	—
FASD8	5/5	Typical	Primary molars, secondary incisors and canines	Typical	—
FASD9	4/4	High	Primary teeth	Typical	—
FASD10	4/4	Slightly heightened	Primary molars, secondary incisors and canines	Typical	—

<sup>a</sup>The Lip-Philtrum Guide (LPhG; Astley, 2014; Hoyme et al., 2015) is a qualification of craniofacial abnormality by lip thickness and philtrum depth on a 5-point scale. The scale ranges from extremely thick/deep (1) to extremely thin/shallow (5), with 3 corresponding to the general population mean. A more detailed description is provided in Appendix A.



Maassen et al., 2017), comprising picture naming, word and nonword repetition, and diadochokinesis (DDK) tasks. The speech perception assessment comprised the auditory discrimination of word and nonword tasks of the Psycholinguistic Assessment of Language Processing in Aphasia (PALPA) Test Battery (Bastiaanse, Bosje, & Visch-Brink, 1995). Although this test is originally designed for adults with aphasia, it has been adapted for children and is widely used in Flanders and the Netherlands to assess children and other disordered populations (e.g., Coppens-Hofman, Terband, Snik, & Maassen, 2016; De Bleser, Faiss, & Schwarz, 1995; Nijland, 2009; Sasisekaran & Luc, 2006; Sasisekaran, Luc, Smyth, & Johnson, 2006; Terband et al., 2011, 2014; Terband, van Zaalén, & Maassen, 2012). General oral motor skills were tested with the oral motor movement assessment (OMMA) from the Dutch Dyspraxia Programme (Erlings-van Deurse, Freriks, Goudt-Bakker, Van der Meulen, & de Vries, 1993). A detailed description of the tests can be found in Appendix A.

The data were collected at the children's school, a speech clinic, or a familiar local community center. All children were given ample time to rest and play between tasks. The ICS-NL questionnaire was completed by one of the children's parents/caretakers. As part of a larger study comprising several additional experimental tasks (Nijssen et al., 2015; Terband & van Brenk, 2015; Terband et al., 2014; van Doornik et al., 2018), data collection in the TD children took place during two 60-min sessions planned in 2 consecutive weeks. For each child with an FASD, one 60- to 90-min session was planned to collect all data (enabling them to take extra time to rest and play if necessary). Regarding the group with FASD, the assessments were administered by the second author, whereas the assessments of TD children were carried out by an independent SLP. The PALPA and the CAI were administered by computer and presented over headphones (Philips SBC HP800). For the CAI, audio was recorded by an omnidirectional externally powered table microphone (Shure 2XU).

### ***Data Processing and Transcription Analyses***

Of the standardized production, perception, and oral motor tasks, the ICS-NL, OMMA, and DDK tasks of the CAI were scored by the second author and an independent SLP, whereas the PALPA was scored automatically by computer. The picture naming, word repetition, and nonword repetition tasks of the CAI were evaluated by a phonetic accuracy and phonological error analysis based on broad phonetic transcription according to the CAI analysis protocol (Maassen et al., 2017). The produced utterances were transcribed and scored in consensus by the first and second authors and an independent transcriber (a Dutch licensed SLP). Point-to-point agreement for the initial transcriptions was 95% for the TD group and 90% for the group with FASD. Transcriptions were analyzed with the Logical International Phonetics Program (Intelligent Hearing Systems, 2012), a computer-

based system that allows transcribed utterances to be analyzed with respect to their phonetic and phonological characteristics.

A comparison of produced and target utterances was conducted at the segmental level. The resulting variables are detailed in Table 3. Analyses concerned the identity of the segments in syllable-initial position<sup>1</sup> and yielded two types of variables: proportions of consonants correct (overall and in the case of the picture naming task also separated out for different developmental complexity levels; this is further explained below) and proportions of substitutions and deletions.

The phonological substitutions were further broken down into phonological features and phonological processes, classified as typical or atypical substitution processes. Young children and children with speech impairment may produce errors that affect entire classes of sounds rather than individual sounds. At particular stages during typical development, children exhibit speech errors that follow patterns based on speech motor skills and phonological knowledge of contrastive characteristics of (categories of) speech sounds. These so-called phonological processes are a normal, natural part of speech development and therefore denoted as typical processes. For Dutch,<sup>2</sup> these typical processes comprise fronting, stopping of fricatives, nasalization, voicing, devoicing, and gliding (Beers, 1995; see also Table 3). A speech profile containing processes that are typical for younger children can be interpreted as speech delay. In contrast, atypical speech processes comprise types of errors that do not usually occur during any stage of speech development and are therefore taken to indicate speech disorder. Backing, abnormal stopping, h-zation, nasalization, dentalization, and lateralization are considered atypical processes for Dutch (Beers, 1995; see also Table 3).

As mentioned above, phonetic accuracy on the picture naming task was also broken down for different developmental stages or levels of complexity to assess phonemic inventories (see Table 3). Beers (1995) developed a system to analyze phonological development, called the Phonological Analysis of Dutch. She found that the phonemic inventory during early childhood speech acquisition develops according to five stages or levels of complexity. These complexity levels are based on the systematic acquisition of phonological features. Individual phonemes can be produced later in development, depending on exposure, but the developmental sequence of phonological contrasts or features is fixed

<sup>1</sup>The focus on consonants in syllable-initial position was motivated by findings that these are the most informative for the assessment of speech production abilities and development in Dutch (Beers, 1995; Coppens-Hofman et al., 2016; Maassen, Terband, van Haften, Diepeveen, & De Swart, 2016; Maassen, van Haften, Diepeveen, De Swart, & Terband, 2015; Maassen et al., 2017; Terband, Coppens-Hofman, Reffeltrath, & Maassen, 2018; see also Ferguson & Farwell, 1975; Stoel-Gammon, 1985, 1987, for English).

<sup>2</sup>A short but comprehensive overview of the phonology of Dutch can be found in, for example, Mennen, Levelt, and Gerrits (2006) and Jonkers, Terband, and Maassen (2014).

**Table 3.** Overview of the variables determined by the segmental comparison of target word and produced utterance.

Phonetic accuracy measures		
PCCI	Proportion of syllable-initial consonants correct	
PCCCI	Proportion of syllable-initial consonant clusters correct	
Phonological error measures		
General		
PSubCI	Proportion of substitutions of syllable-initial consonants	
PDeICl	Proportion of deletions of single syllable-initial consonants	
PRedCLusl	Proportion of reductions of syllable-initial clusters of two consonants	
Phonological features		
PSubPlaceCI	Proportion of substitutions of place of articulation in syllable-initial consonants	
PSubMannerCI	Proportion of substitutions of manner of articulation in syllable-initial consonants	
PSubVoicingCI	Proportion of substitutions of voicing in syllable-initial consonants	
Phonological processes		
PTypSubCI	Proportion of typical processes: substitutions of syllable-initial consonants typical for a speech delay	
Fronting	Consonants made posterior to the alveolar ridge are substituted by another consonant that is made at or in front of the alveolar ridge	
Stopping of fricatives	Fricative or affricate replaced by a plosive	
Denasalization	Replacing a nasal consonant with a homorganic stop	
Voicing	Adding voicing to an unvoiced consonant	
Devoicing	Unvoiced production of a voiced consonant	
Gliding	A liquid replaced with a glide (mostly /j/ or /u/)	
PATypSubCI	Proportion of atypical processes: substitutions of syllable-initial consonants indicative for a speech disorder	
Backing	A labial, alveolar, or dental consonant substituted by a velar /k g ŋ/ or glottal /ʔ/consonant	
Abnormal stopping	Abnormal stops (nonfricative consonant replaced by a plosive)	
H-zation	Replacing a consonant with /h/	
Nasalization	Nasalization of a nonnasal consonant	
Dentalization	Replacing a labial consonant by a coronal	
Lateralization	Replacing a consonant by a lateral /l/	
Developmental complexity measures (phonemic inventory)		
PCC L1CI–L5CI	Proportions of syllable-initial consonants correct at each of the levels of complexity for Dutch, ranging from 1 (least complex) to 5 (most complex; Beers, 1995)	
Level	Feature/contrast	Speech sounds
L1CI	Sonorant, labial, coronal	/p t m n j/
L2CI	Dorsal	/k/
L3CI	Continuant	/f s x h/
L4CI	Front, rounded, (voicing) <sup>a</sup>	/u (b d)/
L5CI	Lateral, rhotic, nasal	/l r/

<sup>a</sup>The age at which the voicing contrast is acquired—/b d/ thereby contrasting with /p t/—is variable and dialect dependent. In general, voicing is a Level 4 contrast, but for some children, it is one of the first, and for some, it is one of the last contrasts they acquire (Beers, 1995; Maassen et al., 2006).

<sup>a</sup>The age at which the voicing contrast is acquired—/b d/ thereby contrasting with /p t/—is variable and dialect dependent. In general, voicing is a Level 4 contrast, but for some children, it is one of the first, and for some, it is one of the last contrasts they acquire (Beers, 1995; Maassen et al., 2006).

(between age categories). A level is considered to be acquired if the speech sounds in that category are correctly produced in 75% of the cases in a representative elicited spontaneous speech sample (for a detailed description of the methodology, see Beers, 1995). In the typical developmental pattern, lower levels of complexity are acquired before higher levels such that, during development, proportions of correct productions tend to be lower at the higher complexity levels. Deviant development can result in a pattern in which a higher level has been acquired (according to the 75%-correct criterion), whereas one or more of the lower levels have not (Maassen, Van der Meulen, & Beers, 2006).

### Statistical Analyses

The level of significance was set at  $p < .05$ , whereas  $p$  values  $< .1$  were qualified as statistical trends. Categorical data were analyzed by means of Pearson's chi-squared tests.

With respect to continuous dependent variables, Shapiro's test of normality, Levene's test of homogeneity of variance, Box's test of homogeneity of covariance, and Mauchly's test of sphericity were carried out where appropriate on all outcome measures before comparing the groups by a series of statistical analyses. These requirements were satisfied for the intelligibility assessment (ICS) and the auditory discrimination and oral motor tasks. In these cases, statistical analysis was done by means of analyses of variance with group as a between-subjects factor, task as a within-subjects factor where appropriate, and age as a covariate. Significant main and interaction effects were further explored by means of univariate tests where appropriate or pairwise comparisons using Fisher's least significant difference test.

For most of the phonetic accuracy and phonological error measures, the results showed that not all requirements of a standard analyses of variance were satisfied, and statistical testing was done by means of a series of generalized

linear mixed models that were adjusted for violations of the assumptions of normality, homogeneity, and sphericity where appropriate (Max & Onghena, 1999; Quené & van den Bergh, 2004).

Regarding the phonetic accuracy and general phonological error measures, generalized linear mixed model analyses were carried out for each outcome measure separately with subject and task as correlated terms, group and task as fixed factors, and age as a random covariate. Significant main and interaction effects were further explored by means of univariate tests where appropriate or pairwise comparisons using Fisher's least significant difference test. For the error measures regarding phonological features, phonological processes, and developmental complexity, single generalized linear mixed model analyses were carried out with subject, task, and type/level of complexity as correlated terms; group, task and type/level of complexity as fixed factors; and age as a covariate.

Correlations between the children's scores on primary outcome measures were calculated separately for both groups using Spearman's  $r$ . As primary outcome measures, we selected the intelligibility judgments (ICS-NL; McLeod et al., 2013), the proportion of syllable-initial consonants correct (PCCI) for the three different speech tasks (CAI; Maassen et al., 2017), the auditory discrimination tasks (PALPA; Bastiaanse et al., 1995), and overall score on the OMMA (Erlings-van Deurse et al., 1993) to keep the number of comparisons feasible. The more conservative Spearman's rather than Pearson's correlation coefficient was used considering the limited sample size. A correction to adjust for multiple statistical tests was not applied as this creates an unacceptably high probability of making a Type II error in analyses with small group sizes (Nakagawa, 2004), and multiple comparisons are accounted for in the interpretation of the results (conform, e.g., Rothman, 1990; van Brenk, Terband, van Lieshout, Lowit, & Maassen, 2013). Rather than focusing on isolated outcome measures, our data analysis and interpretation focused on the patterning of results—on both the group level (FASD vs. TD) and the within-subjects level.

## Results

### *Intelligibility*

The parent/caregiver speech intelligibility judgments (ICS-NL; McLeod et al., 2013) yielded a mean intelligibility score of 4.3 ( $SD = 0.6$ ) for the group of boys with FASD and 4.6 ( $SD = 0.4$ ) for the group of TD children. Statistical analysis (note that age was included as a covariate) revealed the effect of group to be marginally significant,  $F(1, 34) = 4.136, p = .050$ , indicating relatively lower mean intelligibility scores in the group of boys with FASD compared with the TD children.

### *Speech Production Tasks: Phonetic Accuracy Measures*

Data analysis of the speech production tasks featured a layered approach: We first explored the phonetic accuracy

measures and then conducted a series of more in-depth analyses in terms of types of errors, phonological processes, and developmental complexity.

Group-based results of the phonetic accuracy measures on the speech production tasks are presented in Figure 2. With respect to the PCCI, statistical analyses yielded a significant main effect of group,  $F(1, 31) = 30.665, p < .001$ , as well as a significant main effect of task,  $F(2, 50) = 33.133, p < .001$ , and a significant Group  $\times$  Task interaction,  $F(2, 50) = 3.502, p < .05$ . Pairwise comparisons indicated lower proportions of initial consonants correct in the group with FASD as compared with the TD group on all speech tasks (all  $ps < .01$ ). Furthermore, both groups showed lower scores on the nonword repetition task as compared with word repetition and picture naming (all  $ps < .001$ ), whereas performance on the latter two was similar. However, the increase of errors in nonword repetition compared with the other two tasks was larger for the boys with FASD than for the TD children.

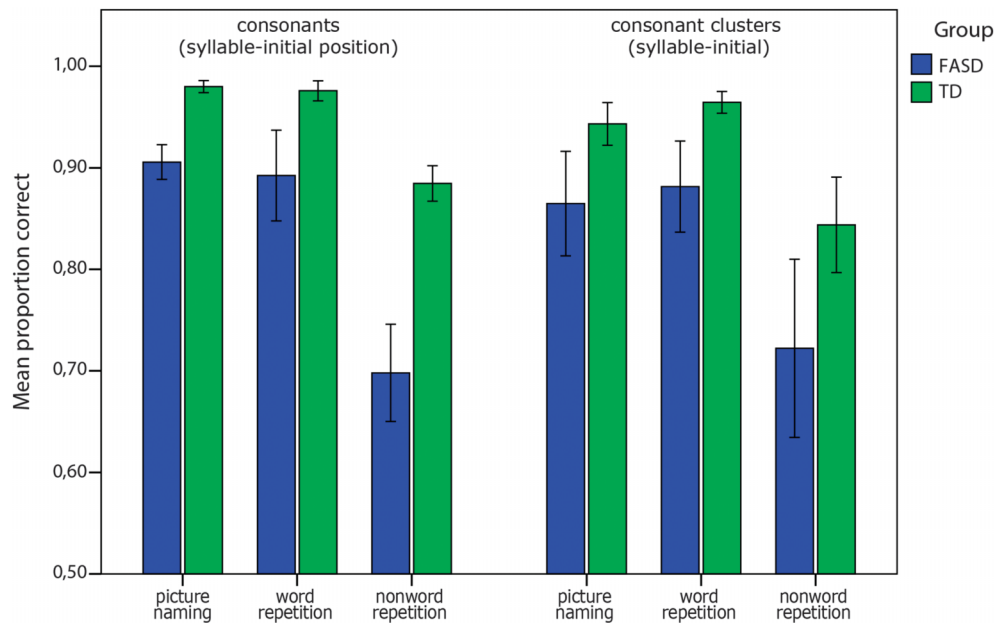
For the proportion of syllable-initial consonant clusters correct (PCCCI), statistical testing revealed a significant main effect of group,  $F(1, 29) = 24.075, p < .001$ , indicating lower scores in the group with FASD as compared with the TD group across speech tasks. The analysis also revealed a significant effect of task,  $F(2, 56) = 8.588, p < .001$ , but no significant Group  $\times$  Task interaction. Pairwise comparisons showed that the effect of lower PCCCI in the group with FASD as compared with the TD group held up for all speech tasks (all  $ps < .01$ ). Furthermore, the boys with FASD showed lower PCCCI scores on the nonword repetition task as compared with word repetition and picture naming (all  $ps < .05$ ), whereas performance on the latter two was similar. The TD children also showed lower scores on the nonword repetition as compared with word repetition ( $p < .05$ ), but the contrasts involving picture naming did not reach significance.

### *Speech Production Tasks: Phonological Error Measures*

Figure 3 presents mean group-based results on the phonological error measures. Statistical testing revealed a significant main effect of group,  $F(1, 40) = 8.931, p < .01$ , for the proportion of deletions of syllable-initial consonants, indicating higher scores in the group with FASD as compared with the TD group across speech tasks. The analysis also revealed a significant effect of task,  $F(2, 98) = 3.343, p < .05$ , but the Group  $\times$  Task interaction was not significant. Pairwise comparisons showed a higher proportion of syllable-initial consonant deletion in the group with FASD as compared with the TD group for word and nonword repetitions (both  $ps < .05$ ) and a trend effect for the picture naming task ( $p = .069$ ). Despite the significant main effect of task, however, the pairwise contrasts only revealed a marginally significant difference between nonword repetition and picture naming for the group with FASD ( $p = .050$ ) and a trend of a difference between nonword repetition and word repetition for the TD group ( $p = .088$ ). All other comparisons did not approach significance.



**Figure 2.** Mean scores per group on the phonetic accuracy measures broken down by speech task. Error bars indicate 1 SE. FASD = fetal alcohol spectrum disorder; TD = typically developing.



With respect to the proportion of reductions of syllable-initial two-consonant clusters, the statistical analysis also revealed a significant main effect of group,  $F(1, 32) = 14.342$ ,  $p < .001$ , and task,  $F(2, 58) = 9.220$ ,  $p < .001$ , but no significant Group  $\times$  Task interaction. Pairwise comparisons showed a higher proportion of cluster reduction in the group with FASD as compared with the TD group for word and nonword repetitions (both  $ps < .05$ ) and a trend effect for the picture naming task ( $p = .088$ ). Furthermore, both groups showed a higher proportion of cluster reductions in the nonword repetition task as compared with word repetition and picture naming (all  $ps < .05$ ), whereas the outcomes on the latter two were similar.

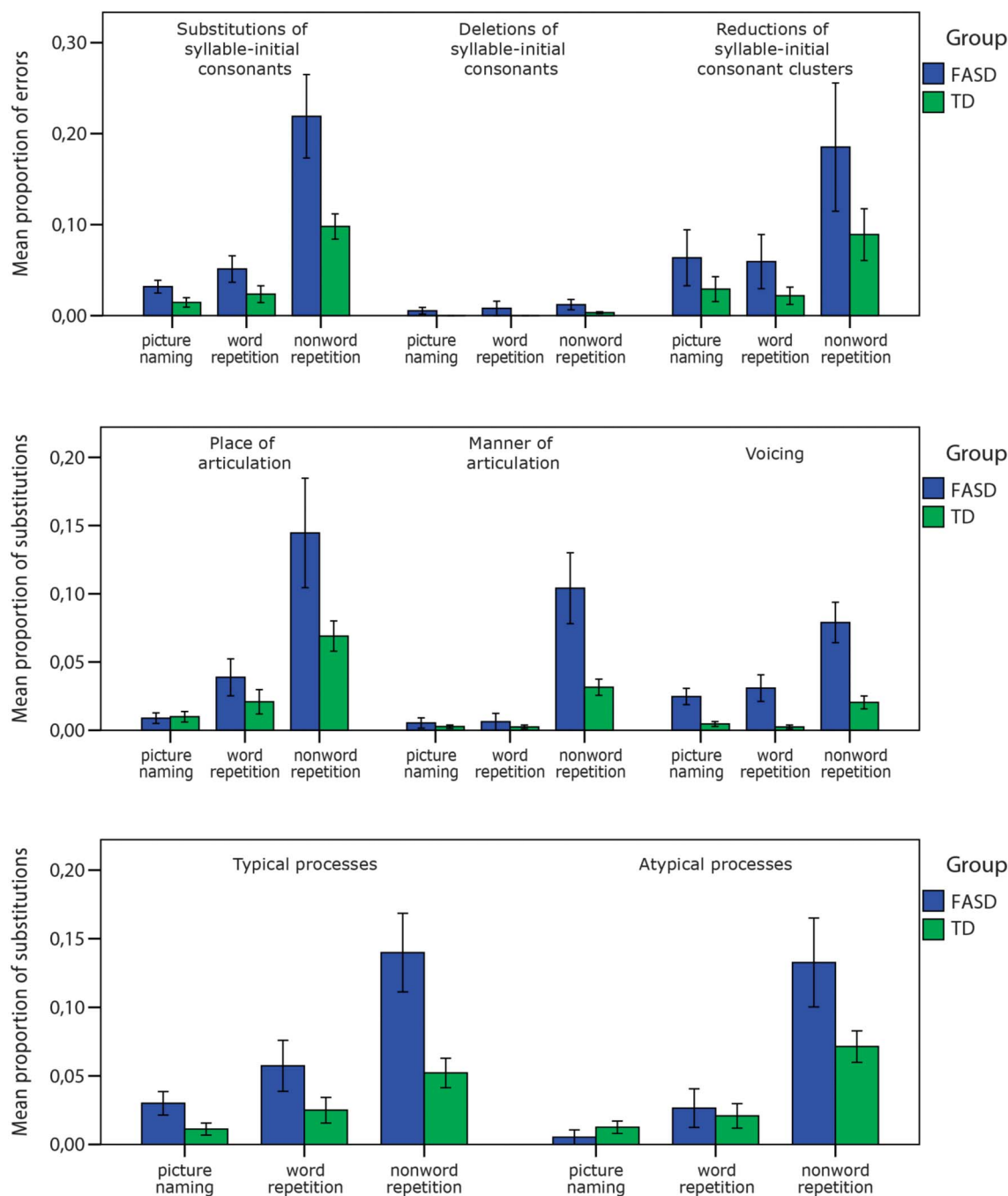
Regarding the proportion of substitutions of syllable-initial consonants, statistical analyses yielded a significant main effect of group,  $F(1, 24) = 12.789$ ,  $p < .01$ , as well as a significant main effect of task,  $F(2, 66) = 60.900$ ,  $p < .001$ , and a significant Group  $\times$  Task interaction,  $F(2, 66) = 9.908$ ,  $p < .001$ . However, pairwise comparisons indicated that the boys with FASD only made more substitutions as compared with the TD children in the nonword repetition task ( $p < .001$ ), whereas in this respect, the groups did not differ on word repetition and picture naming. Comparing between tasks, both groups showed more syllable-initial consonant substitutions in the nonword repetition task as compared with word repetition and picture naming (all  $ps < .001$ ), whereas the outcomes on the latter two were similar. In addition, the increase of the proportion of substitutions in nonword repetition compared with the other two tasks was larger for the boys with FASD than for the TD children.

Further phonological error analyses were conducted to gain more insight into the processes underlying the speech

production problems. We first investigated whether the patterning of the specific types of errors was similar to the error pattern observed in the TD children in terms of phonological features, divided into substitutions of place of articulation, manner of articulation, and voicing (see also Table 3). The statistical analysis revealed a significant main effect of group,  $F(1, 294) = 50.624$ ,  $p < .001$ , as well as significant main effects of task,  $F(2, 294) = 85.399$ ,  $p < .001$ , and type of substitution,  $F(2, 294) = 10.611$ ,  $p < .001$ , as well as Group  $\times$  Task,  $F(2, 294) = 18.486$ ,  $p < .001$ , and Task  $\times$  Type,  $F(4, 294) = 5.924$ ,  $p < .001$ , interactions. The Group  $\times$  Type and Group  $\times$  Task  $\times$  Type interactions were not significant. A series of pairwise comparisons indicated a pattern of results of higher proportions of substitutions in the group with FASD as compared with the TD group for all types of substitutions (all  $ps < .001$ ) and more speech errors involving substitution of place of articulation in comparison with manner of articulation and voicing for both groups (all  $ps < .05$ ). Logically, following the results on the general proportion of substitution measure, pairwise comparisons indicated a larger increase in the proportion of substitutions in nonword repetition compared with the other two tasks for the boys with FASD than for the TD children. Comparing between types, the results showed more substitutions of place of articulation compared with manner of articulation and voicing ( $p < .001$ ) as well as a trend of a difference between manner of articulation and voicing ( $p = .062$ ) for the nonword repetition task across groups. For word repetition, the results revealed a similar difference between place and manner of articulation ( $p < .05$ ).

Subsequently, we investigated whether the patterning of the specific types of errors was similar to the error pattern

**Figure 3.** Mean proportions of different types of errors broken down by speech task and group. Error bars indicate 1 SE. FASD = fetal alcohol spectrum disorder; TD = typically developing.



observed in the TD children in terms of phonological processes. The phonological substitutions thus were divided into typical and atypical processes (see also Table 3). Statistical testing yielded a significant main effect of group,  $F(1, 196) = 24.963, p < .001$ , as well as a significant main effect of task,  $F(2, 196) = 50.668, p < .001$ , and a Group  $\times$  Task,  $F(2, 196) = 8.653, p < .001$ , interaction. The results

furthermore showed a trend of a Group  $\times$  Type interaction,  $F(1, 196) = 3.106, p < .080$ . Pairwise comparisons showed more substitutions for the boys with FASD compared with the TD children (both  $ps < .05$ ) with a relatively larger increase of the number of typical substitutions for the group with FASD in comparison with the TD children. Logically, pairwise comparisons again indicated a larger increase in the proportion of

substitutions in nonword repetition compared with the other two tasks for the boys with FASD than for the TD children.

An overview of the processes per group as well as per child with an FASD is presented in Appendix B. The most eye-catching aspect is the high dispersion of error types with respect to both phonological features and phonological processes. Apart from participant FASD8 (who did not complete the nonword repetition task and whose errors on picture naming and word repetition comprised predominantly deletions), the boys with FASD exhibited between 6 and 10 of 12 different processes. Although individual results showed there was variation among the boys with FASD, the results did not reveal clear idiosyncratic error patterns. On the group level, the results indicated that fronting was the most common process in both groups. Furthermore, the results showed relatively high proportions of denasalization, voicing, and devoicing for the boys with FASD compared with the TD children. A detailed examination of the distribution of these errors among the boys with FASD revealed that denasalization did not occur in the two boys with a normal palate (FASD5 and FASD8) and did occur in all the boys with FASD who featured a heightened palate (see Table 2 and Appendix B). No patterns were observed with respect to other error types, and the results also did not reveal any pattern in type or number of errors that was specific to the three participants with hearing loss (FASD2, FASD3, and FASD8).

### ***Speech Production Tasks: Developmental Complexity Measures (Phonemic Inventory)***

Our next query was to compare the phonemic inventories of the boys with FASD with those of the TD group on the pattern described for typical speech acquisition. Figure 4 shows the mean proportions of syllable-initial consonants correct for each of the complexity levels for the group of boys with FASD compared with the TD group, whereas individual values for the boys with FASD are presented in Appendix B. (Note that phonemic inventories were evaluated on the picture naming task only.) The statistical analysis revealed significant main effects of group,  $F(1, 165) = 45.978, p < .001$ , and level,  $F(4, 165) = 10.957, p < .001$ , as well as a trend of a Group  $\times$  Level,  $F(4, 489) = 2.470, p = .064$ , interaction effect. As the results on the general measure PCCI already indicated, the boys with FASD produced lower proportions of consonants correct than the TD children. Pairwise comparisons indicated that this between-group difference held up for all categories of complexity except Level 2 consonants (for all other levels,  $p < .05$ ). Furthermore, pairwise comparisons indicated a significant main effect of level for both groups (both  $ps < .01$ ). For the group with FASD, the mean proportion correct on Level 5 consonants was significantly lower than those on all the other levels, and the proportion correct on Level 3 consonants was also significantly lower than on Levels 1 and 2 (all  $ps < .05$ ). For the TD group, only the proportion of consonants correct on Level 5 was significantly lower than all other levels (all  $ps < .01$ ); no other contrasts were different from each other.

On the group level, the high mean values (all above the 75%-correct criterion; see Figure 4) indicate that, overall, the phonological repertoire is complete. The values per participant (see Appendix B), however, show that not all boys with FASD reached the 75%-correct criterion at all complexity levels. Furthermore, across the board, the results show a tendency of a decline in the proportion of consonants correct with increasing complexity but simultaneously there is a tendency for Level 2 (/k/) and Level 4 (/v b d/) consonants to be more frequently produced correctly than consonants at the other levels (with the exception of FASD8).

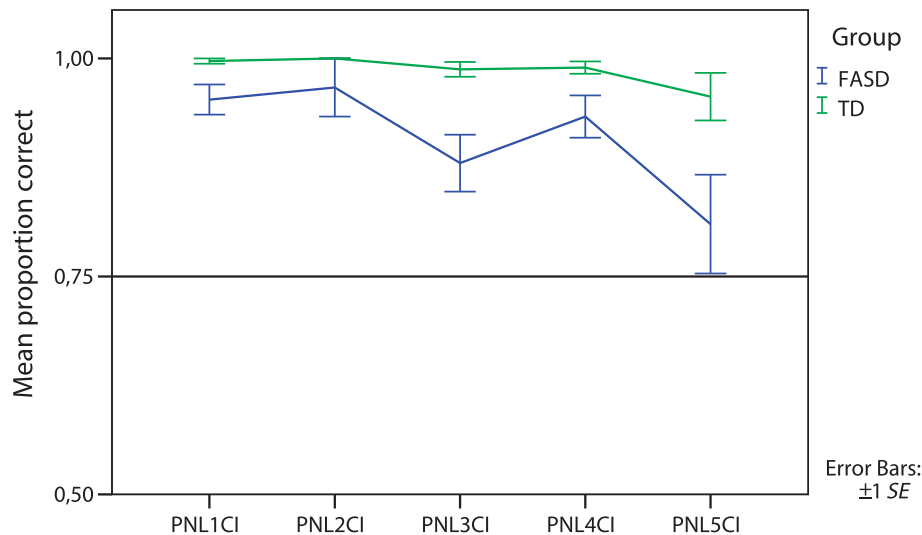
### ***Auditory Discrimination and Oral Motor Tasks***

Auditory discrimination and oral motor abilities were measured on a functional level to investigate their potential role in the speech impairment. Table 4 presents the group results of the auditory discrimination assessment and OMMA, whereas the results of the DDK assessment are presented in Table 5. Statistical analyses revealed significant main effects of group for the auditory discrimination,  $F(1, 29) = 5.440, p = .027$ , OMMA,  $F(1, 15) = 44.737, p < .001$ , and DDK score ( $\chi^2 = 6.235, p = .013$ ) and judgment ( $\chi^2 = 15.079, p = .002$ ), indicating that the scores of the boys with FASD across all three tasks were lower than those of the TD children. The apparent interaction of PALPA Task (words vs. nonwords)  $\times$  Group did not reach significance,  $F(1, 29) = 2.562, p = .120$ , and also no significant OMMA Task (isolation, sequential, and sequential fast)  $\times$  Group interaction was observed. A detailed examination of the individual results among the boys with FASD did not reveal any patterns regarding the participants with hearing loss (FASD2, FASD3, and FASD8) that might indicate a relation between hearing acuity and auditory discrimination and oral motor abilities (see Table 1 and Appendix B).

### ***Correlations Between Intelligibility, Speech Production, Auditory Discrimination, and Oral Motor Tasks***

To gain further insight into the processes underlying the speech production problems, we calculated the correlations between the scores on the intelligibility and speech production assessments on the one hand (i.e., speech intelligibility, PCCI picture naming, and word and nonword repetitions) and the intelligibility, oral motor movement, and auditory discrimination assessments (i.e., speech intelligibility, word and nonword auditory discrimination, and overall score on oral motor movements) on the other hand, separately for both groups. The correlation matrix is presented in Table 6. Results revealed very different patterns for the two groups. For the TD group, auditory discrimination abilities were positively correlated with the PCCI on the speech production tasks, whereas none of the measures was correlated to the intelligibility judgments. Interestingly, the results of the group with FASD showed a pattern in which oral motor performance was strongly correlated (positively) with PCCI picture naming and word repetition

**Figure 4.** Mean proportions of correctly produced consonants according to developmental levels of complexity for Dutch (Beers, 1995) on the picture naming task (Computer Articulation Instrument; Maassen et al., in press) for the children with fetal alcohol spectrum disorders (FASD) compared with the typically developing (TD) children. The reference line denotes the 75%-correct criterion above which a speech sound category is considered to be acquired. Error bars indicate 1 SE. PN = picture naming; L1CI–L5CI: Level of complexity 1–5 of consonants in syllable-initial position.



but not with PCCI nonword repetition. PCCI nonword repetition, however, did show a strong positive correlation with intelligibility, and also PCCI word repetition and auditory discrimination of words and nonwords were positively correlated with intelligibility.

### Summary of Findings

This study examined the phonological and speech motor characteristics in boys with FASD. In summary, the results showed that the boys with FASD were less intelligible and made more consonantal errors compared with the TD children. Comparing between speech tasks, both groups showed lower scores on the nonword repetition task as compared with word repetition and picture naming, whereas performance on the latter two was similar, but this effect was stronger in the group with FASD. In addition, the group of boys with FASD also scored lower than the TD group on auditory discrimination and oral motor tasks and showed a different pattern of correlations between

auditory discrimination and oral motor abilities, phonetic accuracy (PCCI), and intelligibility compared with the TD children.

The specifics of the speech errors were further investigated in a layered manner. First, we analyzed the occurrence of substitutions, deletions, and cluster reductions and found higher error rates in the speech of the boys with FASD compared with the TD children for all three types. Furthermore, the results showed a general pattern of higher proportions of all three types of speech errors in the nonword repetition task as compared with word repetition and picture naming, whereas the outcomes on the latter two were similar. However, for substitutions, the boys with FASD showed a larger increase of errors in nonword repetition compared with the TD children, whereas the increase was similar across groups for deletions and cluster reductions.

The substitutions were then further analyzed and broken down in terms of phonological features (divided into substitutions of place of articulation, manner of articulation, and voicing) and phonological processes (divided

**Table 4.** Mean (SD) scores per group on the auditory discrimination and oral motor movement assessment subtests.

Group	Age (years)	Auditory discrimination (PALPA) <sup>a</sup>		Oral motor movement assessment <sup>b</sup>	
		Words, % correct (SD)	Nonwords, % correct (SD)	Isolation, sequential, sequential fast, % correct (SD)	Overall, % correct (SD)
FASD	7.2 (1.9)	74.9 (9.6)	78.3 (18.1)	88.1 (12.9), 80.3 (11.6), 68.0 (10.3)	82.0 (10.4)
TD	5.6 (1.4)	83.7 (15.0)	78.9 (14.4)	97.0 (4.1), 93.5 (6.4), 88.8 (12.5)	94.4 (5.6)

Note. FASD = fetal alcohol spectrum disorder; TD = typical development.

<sup>a</sup>Psycholinguistic Assessment of Language Processing in Aphasia (Bastiaanse et al., 1995). <sup>b</sup>Erlings-van Deurse et al., 1993.



**Table 5.** Performance on diadochokinesis assessment (DDK; [pataka]) of the Computer Articulation Instrument (CAI; Maassen et al., in press) per group by means of the numbers of participants who scored in the respective categories.

Assessment outcome	FASD (n = 9)	TD (n = 23)
DDK score		
[pataka] could be produced	4	20
[pataka] could not be produced	5	3
DDK judgment		
Perfect	0	12
[pataka] in sequence in normal rate, but no acceleration	1	7
[pataka] in sequence incorrect ([t] or [k] could not be pronounced), but speeding up on two different consonants ([pata], [taka]) was possible	7	4
No fluent [pataka], not in sequence	1	0
No [pataka] production either in isolation or in a sequence of two	0	0

Note. FASD = fetal alcohol spectrum disorder; TD = typical development.

into typical and atypical substitutions). The results of the phonological feature analysis revealed error patterns that were very similar across groups. Both groups tended to produce more speech errors involving the substitution of place of articulation compared with manner of articulation and voicing, especially during nonword repetition, but no interactions involving group and type were found. The analysis of phonological processes, on the other hand, did reveal such an interaction, and the results showed that, in the group with FASD, the difference in the number of typical substitutions compared with the TD children was larger than the difference in the number of atypical substitutions compared with the TD children. This higher number of errors comprised notably the processes of denasalization, voicing, and devoicing, but both group-based and individual results showed a high dispersion of error types. The results did not reveal idiosyncratic error patterns and any error pattern that was specific to participants with or without hearing loss. In other words, no core of specific speech errors that were typical for boys with FASD could be identified. The results, however, do suggest that there might be specific speech errors that are related to specific craniofacial structural defects. A detailed comparison of the craniofacial and orofacial anatomical characteristics

(see Table 2) with error types (see Appendix B) revealed a pattern in which the boys with FASD who featured a heightened palate all showed denasalization, whereas the two boys with a normal palate (FASD5 and FASD8) did not make any errors involving denasalization. This pattern suggests that these denasalization errors are not phonological substitutions but rather result from the structural defect of a heightened palate.

Finally, we compared the phonemic inventories of the boys with FASD with those of our TD group and differentiated the speech errors on the picture naming task according to the levels of complexity of Beers' Phonological Analysis of Dutch (Beers, 1995). The results showed that, for all levels, the proportions correct were above the 75%-correct criterion, indicating that, overall, the phonological repertoires were complete (for both the TD group and the group with FASD). However, it should be noted that not all boys with FASD reached the 75%-correct criterion at all complexity levels and also, at the group level, the results showed interesting differences between levels. Across the board, the results showed a tendency of a decline in the PCCI with an increasing complexity similar to the TD children. At the same time, the results indicated that the boys with FASD show a dip for complexity level 3 (/f s x h/)

**Table 6.** Spearman's correlations between parent/caretaker intelligibility judgments (ICS-NL; McLeod et al., 2013), proportion of syllable-initial consonants correct (PCCI) for the three different speech tasks (CAI; Maassen et al., in press) and auditory discrimination tasks (PALPA; Bastiaanse et al., 1995), and overall score on the oral motor movement assessment (OMMA; Erlings-van Deurse et al., 1993) for the group of boys with FASD and the comparison group of typically developing (TD) children.

Task	FASD				TD			
	ICS-NL	PALPA words	PALPA nonwords	OMMA overall	ICS-NL	PALPA words	PALPA nonwords	OMMA overall
ICS-NL	—	.73*	.78*	.38	—	.08	.04	-.02
PCCI picture naming	.29	.25	.16	.86**	.02	.56**	.48*	.04
PCCI word repetition	.74*	.45	.39	.76**	-.02	.53**	.49*	-.22
PCCI nonword repetition	.93**	.51	.53	.22	-.08	.64**	.67**	.29

Note. CAI = Computer Articulation Instrument; PALPA = Psycholinguistic Assessment of Language Processing in Aphasia; FASD = fetal alcohol spectrum disorder; ICS-NL = Intelligibility in Context Scale.

\* $p < .05$ . \*\* $p < .01$ .

and, at this level, they produced more errors than would be expected based on their performance on the adjacent levels of complexity.

## Discussion

### *Is Speech Development in FASD Disordered or Delayed?*

To establish whether speech development in boys with FASD is delayed or (also) deviant, we investigated two dimensions of speech development: the order in which speech sounds are typically acquired and the way speech sounds are typically produced during the process of acquisition. The order of acquisition was analyzed based on Beers' (1995) Phonological Analysis of Dutch in which the phonemic inventory is divided into five developmental levels of complexity. The way speech sounds are acquired was investigated by analyzing the type of errors that were made in terms of phonological processes divided into typical (which are indicative for a speech delay) and atypical (which are indicative for a speech disorder) substitution processes.

The comparison of phonological processes showed that the group with FASD predominantly produced more typical substitutions compared with the TD children. In picture naming and word repetition, the boys with FASD did not produce more atypical substitutions than the TD children, and their number did not increase disproportionately in the nonword repetition task. In other words, the lower proportion of consonants correct in the boys with FASD compared with the TD children did not stem from an increase in atypical substitutions but consisted mainly of processes typical for younger children. On this dimension, the results indicate that speech impairment in FASD involves developmental delay.

The analysis of the order of acquisition of phonemic inventories, however, suggests that this is not the whole story. An incomplete inventory typically results in a pattern in which the higher levels are produced at a lower percentage accuracy, whereas an overall lower percentage accuracy across complexity levels indicates inconsistency of production (Thoonen, Maassen, Gabreels, & Schreuder, 1994). The present group-level results indicated that, in our sample of boys with FASD, the phonological repertoire was complete as well as a general tendency of a decline in the proportion of consonants correct with increasing complexity, a pattern that is compatible with speech delay. Comparing the different developmental levels of complexity, however, the results also show a tendency for Level 2 (/k/) and Level 4 (/v b d/) consonants to be more frequently produced correctly by the boys with FASD than consonants at the other levels, meaning that a subset of errors was made irrespective of phonological complexity. This pattern in which (some) lower levels are outperformed by (some) higher levels is not observed in the TD group and indicates that speech development in FASD is not only delayed but shows signs of deviance in the acquisition of phonological features as well.

### *Possible Underlying Mechanisms*

The results showed that the boys with FASD scored lower compared with the TD children on auditory discrimination. These lower auditory discrimination scores in the group with FASD could not be related to the presence or absence of hearing loss. The results did not reveal any statistically significant differences between the auditory discrimination of words and nonwords, and the correlational analysis revealed correlations with intelligibility of both auditory word and nonword discrimination. On first account, these results suggest that a functional deficit in the auditory discrimination of speech sounds plays a role in the speech impairment in FASD. However, the results of the boys with FASD did not show statistically significant correlations between auditory discrimination and PCCIs on the speech production tasks. Poor auditory discrimination thus cannot be the only mechanism at work, and the question arises why auditory discrimination would be correlated with intelligibility but not with PCCI measures.

With respect to word and nonword repetitions, this might be partly due to lack of statistical power as the *r* values would be indicative for moderate effect sizes, but the correlations fail to reach significance. In addition, the different pattern of correlations between the group with FASD and the TD group reflects that, in the group with FASD, both auditory and motor functions more equally underlie the results on the speech production tasks, as compared with only auditory functions in the TD group. Especially in the production of words (picture naming and word repetition), it can be hypothesized that the quality of word-form storage is not the primary difficulty but that the executive motor functions are instead. The comparisons between speech tasks showed that the boys with FASD performed disproportionately worse than the TD group in the nonword repetition task compared with word repetition and picture naming, but we did not find any differences between auditory word and nonword discrimination. The group with FASD also did not show a significant correlation between the performance on nonword discrimination and nonword repetition.

The question thus arises: What could be responsible for the disproportionate increase in speech errors of the boys with FASD during nonword repetition? Besides the lower scores on auditory discrimination, our results also showed that the boys with FASD scored lower compared with the TD children on general speech motor and oral motor abilities, both on the maximum performance DDK task and on the OMMA. In addition, oral motor movement performance was strongly correlated with PCCI picture naming and word repetition in the group with FASD, indicating that oral motor abilities are playing a role as well. A closer look at the individual functional tasks of the OMMA (a description is provided in Appendix A) in the group with FASD revealed that all children except one (FASD4) had problems with tongue movements and that tongue control was the only aspect that caused problems (with the exception of FASD2, who also showed reduced lip strength). Corroborating evidence for a specific oral

motor deficit involving tongue control comes from our finding that the boys with FASD experienced difficulties in particularly producing Level 3 (/f s x h/) and Level 5 (/l r/) consonants—the categories that contain the speech sounds that rely the most on tongue control for Dutch. (For example, the fricatives /s, x/ require more tongue control as compared with their plosive Level 1 [/t/] and Level 2 [/k/] place-of-articulation counterparts.) From these results, we can conclude that a specific oral motor deficit, that is, problems with tongue control, plays an important role in the speech impairment in boys with FASD. However, because we did not find a correlation between oral motor performance and PCCI nonword repetition, this still cannot be the final story.

The task of nonword repetition poses special demands on the speech perception and production system as the speaker cannot make use of the lexicon and stored word forms. Two routes are possible (e.g., Maassen & Terband, 2015). If the speaker is able to analyze the phonological structure of the nonword, he or she can address the phonological encoding system and select and sequence the linguistic/symbolic units that constitute the nonword. In principle, the subsequent stages of motor planning, programming, and execution could then advance the same as in picture naming and word repetition. However, although the nonword stimuli feature syllable structures similar to the stimuli of the picture naming and word repetition tasks, the nonwords are composed of syllables that do not exist as words in Dutch, and there might be frequency effects of syllables and syllable combinations that still play a role (cf. Mousikou & Rastle, 2015). The second route is needed if the speaker is not able to analyze the phonological structure of the nonword. In this case, nonword repetition is similar to the imitation of nonspeech sounds, and the motor planning system has to be addressed directly. Such imitation relies heavily on the speaker's internal models, first to derive sensory and articulatory goals from the auditory information and, subsequently, to guide motor programming and self-monitoring. To sum up, besides auditory processing and phonological working memory, nonword repetition also poses special demands on the motor planning and motor programming parts of the speech production chain. These extra demands compared with word repetition and picture naming differ depending on the route followed to produce the nonword utterance. This suggests that the underlying deficits are not perceptual but output based and that, indeed, weak motor planning and programming underlie the speech difficulties. However, this cannot be definitively verified based on the data collected in this study. To help pinpoint which processes are responsible for the disproportionate error increase during nonword repetition, future studies could, for example, focus on stimulus length, and syllable structure and frequency effects or on consistency in repeated productions of words and nonwords.

In summary, the present results do not implicate a single subsystem that is responsible for the speech impairment in boys with FASD. Rather, deficits in multiple subsystems, namely, craniofacial structure (heightened palate), auditory discrimination, oral motor control (specifically involving

the tongue), and speech motor planning/programming, all appear to be playing a role. Furthermore, the subsystems responsible for speech impairments in children with FASD will likely differ for each child as not all children in this study had hearing impairment, not all had high-arched palates, not all had difficulty with tongue movements, and not all showed deviated phonological development. Further research is necessary to further unravel how these different subsystems are involved and how they interact in speech production and development in FASD. Recent neuroimaging studies found a decreased surface/volume of the cerebellum and basal ganglia and a less myelinated corpus callosum (Donald et al., 2015; Moore, Migliorini, Infante, & Riley, 2014; Norman et al., 2009) as well as decreased activation in Broca's area in combination with increased activation of the dorsal pathway and cerebellar regions during attention and verbal working memory tasks in children with FASD as compared with TD children (Diwadkar et al., 2013; O'Conaill et al., 2015). It is suggested that, in FASD, processes that are (partly) subserved by the basal ganglia and the cerebellum fall short, especially when task demands increase. In the speech production chain, this implicates sequencing and sensory motor integration that underlie motor planning and motor programming (e.g., Bohland, Bullock, & Guenther, 2010; Guenther, Ghosh, & Tourville, 2006; Guenther & Perkell, 2004). To further specify the mechanisms that underlie speech impairment in FASD, future studies that investigate the role of sequencing and sensory motor integration in connection with speech output measures are warranted.

### *Limitations and Suggestions for Future Studies*

Although the results are consistent among the children who participated in this study, it has to be taken into account that the group of boys with FASD was relatively small in numbers. To further test the strength of the results found in this study, future studies should include a larger sample size.

Furthermore, future studies should be expanded to include also girls with FASD. There are clear indications of gender-based differences in the adverse effects of prenatal alcohol exposure on some childhood developmental outcomes, but not all adhere to this pattern (Abel & Hannigan, 1995; Griesler & Kandel, 1998; Herman, Acosta, & Chang, 2007; O'Connor, 2001; Pfander, Liebig, & Feldmann, 2014; Rasmussen, Becker, McLennan, Urchuk, & Andrew, 2011; Sokol et al., 1986; Sood et al., 2001; Terasaki, Gomez, & Schwarz, 2016; Willoughby, Sheard, Nash, & Rovet, 2008). Animal models have shown large differences in the detrimental effects of prenatal alcohol exposure between males and females, indicating that particularly males are vulnerable (Tunc-Ozcan, Ullmann, Shukla, & Redei, 2013). Similarly, several studies have reported gender-related differences with respect to FASD in humans. Some studies have found a higher occurrence of FASD in males as compared with females (e.g., Astley, 2010; May et al., 2007; May, Hymbaugh, Aase, & Samet, 1983; Thanh, Jonsson, Salmon, & Sebastianski, 2014), but it should be noted that

most prevalence studies did not find evidence of differences between males and females (e.g., Fox et al., 2015; May et al., 2014). With respect to the clinical characteristics, there is a growing body of evidence of gender-based differences in the adverse effects of prenatal alcohol exposure (Abel & Hannigan, 1995; Griesler & Kandel, 1998; Herman et al., 2007; O'Connor, 2001; Pfinder et al., 2014; Rasmussen et al., 2011; Sokol et al., 1986; Sood et al., 2001; Terasaki et al., 2016; Willoughby et al., 2008). In general, girls with FASD have been found to exhibit more deficits in social skills (Rasmussen et al., 2011), whereas FASD in boys have been found to involve more cognitive functional deficits such as increased attention deficits (Herman et al., 2007) and reduced accuracy in processing visual stimuli (Paolozza, Munn, Munoz, & Reynolds, 2015). Also in this respect, however, the literature is inconclusive. For example, no effects of gender were found in verbal learning and verbal and spatial recall in children with FASD relative to TD children (Willoughby et al., 2008). Although the evidence is converging toward infant and early childhood developmental outcomes of males prenatally exposed to alcohol being more highly impaired compared with females, at present, "the true scope of sex differences in vulnerability is unknown" (DiPietro & Voegtline, 2017, p. 4). Whether the results found for boys in this study hold up for girls or whether there are gender-related differences in speech and speech motor development in children with FASD is a thus question that warrants further research.

The aim of this study was to profile and characterize speech impairment in FASD. Although this study featured a comprehensive test battery, a multitude of aspects and characteristics of speech production remain to be investigated. For example, this study focused on consonants in syllable-initial position, and future studies could also investigate the production of consonants in other syllabic positions and the production of vowels as well as syllabic error characteristics and processes (e.g., proportions of syllable structures correct and phonological assimilation processes). Given the specific characteristics that are typical for children with FASD, including craniofacial abnormalities and a heightened palate, these may not yield findings similar to past studies of children with different diagnoses. Furthermore, our test battery did not feature a detailed assessment of hearing acuity and type of hearing loss. It is well known that even mild hearing loss can affect children's speech language development negatively (e.g., Briscoe, Bishop, & Norbury, 2001; Crowe & McLeod, 2014; Moeller et al., 2010). Although our present results did not reveal any relation between hearing loss and the auditory discrimination, oral motor, and phonological error measures, it cannot be ruled out that it did play a role in the children's speech motor and phonological development. If possible, future studies should include such detailed assessments to investigate the possible relation of type and severity of hearing loss with the speech profile in children with FASD.

Another limitation lies in the cross-sectional design of the current study. Prospective longitudinal designs focusing on developmental trajectories on fine-grained measures are

needed to fully understand the process of speech motor acquisition in FASD. To establish causal relations would require controlled intervention studies with long-term follow-up measures.

## Conclusions

FASD are highly prevalent in comparison with other congenital syndromes, and the vast majority of the cases involve speech impairment. Yet, to date, the specific characteristics and underlying mechanisms of the speech production problems have not been described in detail, and no dedicated treatment plans have been developed for this population. It is well known that communication disorders affect social competence and that children with poor verbal communication skills often suffer social-emotional and behavioral problems (e.g., Conti-Ramsden & Botting, 2004; Van Daal, Verhoeven, & Van Balkom, 2007), threatening academic skills and occupational opportunities into adulthood (e.g., Felsenfeld, Broen, & McGue, 1994). Moreover, childhood communication disorders have been found to increase the risk of later-life behavioral and psychiatric disorders (e.g., Beitchman et al., 2001; Hinshaw, 1992). Especially for children with FASD, who already face an array of challenges on a wide range of very different areas (i.e., familial, social, socio-economical, cognitive, anatomical), the development of effective treatment methods is of crucial importance—not only to limit the burden of yet another issue but also to confine the negative, catalyzing influence of speech impairment on their other problems.

Effective and efficient intervention requires treatment programs tailored to the specific profile and underlying mechanisms of the speech production problems. By investigating commonalities and individual differences in phonological and speech motor development as compared with TD children, this study aimed to profile and characterize speech impairment in FASD. The results showed that the boys with FASD were less intelligible and made more consonantal errors compared with the TD children. The boys with FASD also showed reduced auditory discrimination and oral motor abilities as well as a deviant pattern of correlations between speech, oral motor, and auditory abilities compared with the TD children. Regarding the type of speech errors, no core of consonantal errors typical for boys with FASD could be identified. The error profile showed strong similarities with those occurring during earlier stages of normal development. However, we also found that a subset of errors was made irrespective of phonological complexity. Furthermore, only the boys with FASD who featured a heightened palate made denasalization errors, indicating that these errors are not phonological substitutions but rather result from the structural deficit. Together, these results indicate that speech development in FASD is both delayed and deviant. Speech impairment in boys with FASD should thus be approached as a complex disorder rather than a developmental delay.

Regarding possible underlying mechanisms, the present findings indicate that the speech impairment in boys



with FASD results from a combination of deficits in multiple subsystems. Furthermore, the exact constellation of subsystems responsible is likely to differ for each child with an FASD. Besides indications of problems with speech motor planning/programming, the boys with FASD in this study showed reduced abilities in the auditory discrimination of speech sounds and reduced oral motor abilities, in particular, tongue control. In addition, individual-specific structural deficits and hearing disorders may play a role. Although these problems might not provide a definitive explanation of the underlying mechanisms, they are all important aspects that influence speech development in FASD and should be taken into account in the design and administration of treatment programs.

As mentioned in the introduction, it has been well established that children with FASD experience difficulties in processing (new) information and learning. This means they need a lot of repetitive practice, not only to acquire new skills but also to become able to implement newly acquired skills into spontaneous speech. Few SLPs are aware of the specific symptoms of FASD and the concomitant problems that make the acquisition of speech and language even more difficult. To improve the speech skills in children with FASD, and thereby reduce the direct and indirect impacts of speech impairment on this already predisposed population, requires long-term dedicated treatment that is tailored to the individual profile under the guidance of SLPs who are trained in working with these children.

## Acknowledgments

This work was supported by the Netherlands Organization for Scientific Research (NWO-VENI Grant 275-89-016; Hayo Terband) and the Dutch Rehabilitation Fund (Grant 2011/0165-060; Ben Maassen and Hayo Terband). The authors gratefully thank the participants and their parents/caretakers for their time and effort as well as Frits van Brenk and two anonymous reviewers for their constructive comments on earlier versions of this article.

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## Appendix A

### Description of the Standardized Speech Production and Perception, and Oral Motor Tasks

Task/assessment	Description
London Postal History Group (LPhG)	The LPhG (Astley, 2014; Hoyme et al., 2015) is a qualification of craniofacial abnormality by lip thickness and philtrum depth based on five pictures of lips and philtrums divided by race (White and African American). The pictures reflect the full range of lip thickness and philtrum depth on a 5-point scale centered around the general population mean. The extreme ends are Ranks 1 and 5, with 1 denoting extremely thick/deep and 5 denoting extremely thin/shallow.
Intelligibility (ICS)	The ICS (McLeod et al., 2013) is a quick parent report measure of children's intelligibility. The seven-item questionnaire rates the degree to which children's speech is understood by different communication partners (parents, immediate family, extended family, friends, acquaintances, teachers, and strangers) on a 5-point scale. A higher score denotes better intelligibility.
Auditory discrimination (PALPA)	Auditory discrimination task from the Dutch translation of the PALPA (Bastiaanse et al., 1995) adapted for children. Score is percentage correct.
Words	36 pairs of CVC words that were either the same (18 pairs), differed on one consonant (initial or final; 12 pairs), or were metatheses of each other (six pairs, e.g., "lor" vs. "rol").
Nonwords	36 pairs of CVC nonwords that were either the same (18 pairs), differed on one consonant (initial or final; 12 pairs), or were metatheses of each other (six pairs, e.g., "tus" vs. "sut").
Diadochokinesis (CAI)	Maximum performance task using utterances of [pataka]. The children were first asked to produce "pataka" once, and when they succeeded, they were asked to produce "pataka" in a sequence of several repetitions of "pataka." After that, the children were asked to speed up while producing a sequence of "pataka." This task is administered with the CAI (Maassen et al., in press).
Professional Teaching Knowledge (PTK) score	1 = [pataka] could be produced; 0 = [pataka] could not be produced.
PTK judgment	4 = perfect; 3 = [pataka] in sequence in normal rate, but no acceleration; 2 = [pataka] in sequence incorrect ([t] or [k] could not be pronounced), but speeding up on two different consonants ([pata], [taka]) was possible; 1 = no fluent [pataka], not in sequence; 0 = no [pataka] production either in isolation or in a sequence of two.
Oral motor movement assessment	Oral motor assessment from the Dutch Dyspraxia Program (Erlings-van Deurse et al., 1993). Percentage of correctly executed oral motor tasks.
Isolation	Positioning of the lips in rest; lip protrusion; lip spreading; stick out the tongue; move the tongue to corners of the mouth; move tongue up; move tongue down; click with the tongue.
Sequential	Lips protrude and spread; move tongue left and right; move tongue up and down; open and close the jaw; check whether velum closes when blowing; check whether velum closes when sucking.
Seq. fast	Same tasks as sequential but in high tempo.
Picture naming (60 words, CAI)	This task consists of 60 images depicting 50 words with different consonants, consonant clusters, and vowels at various positions (initial, medial, and final) and 10 words with complex consonant patterns. This task is administered with the CAI (Maassen et al., in press).
Word repetition (10 words, CAI)	Repetition task using the same 10 words with complex consonant patterns as in picture naming. The words were presented through headphones, and the children were asked to repeat them. This task is administered with the CAI (Maassen et al., in press).
Nonword repetition (33 nonwords, CAI)	Same task as word repetition, using 33 multisyllabic nonword stimuli consisting of syllables that do not exist as words in Dutch. The first 23 nonwords have syllable structures similar to the multisyllabic stimuli of the picture naming task, whereas the last 10 feature complex consonant patterns resembling the stimuli in the word repetition task. This task is administered with the CAI (Maassen et al., in press), similar to word repetition.

*Note.* London Postal History Group = LPhG; ICS = Intelligibility in Context Scale; PALPA = Psycholinguistic Assessment of Language Processing in Aphasia; CVC = consonant–vowel–consonant; CAI = Computer Articulation Instrument, Professional Teaching Knowledge = PTK. Reprinted from *Journal of Communication Disorders*, 51, Hayo Terband, Frits van Brenk, and Anniek van Doornik-van der Zee, "Auditory feedback perturbation in children with developmental speech sound disorders," pp. 64–77, Copyright © 2014, with permission from Elsevier.

## Appendix B

Individual Results for the Boys With FASD on Selected Phonological Error Measures, Auditory Discrimination, and Oral Motor Skills Accompanied by Groups' Means (SD) for the Boys With FASD and the Typically Developing Children (Control)

Outcome measure	FASD1	FASD2	FASD3	FASD4	FASD5	FASD6	FASD7	FASD8	FASD9	FASD10	M (SD) FASD	M (SD) Control
<b>Phonological features<sup>a</sup>: picture naming, word, and nonword repetition tasks combined</b>												
PSubPlaceCI	.045	.105	.112	.000	.066	.009	.031	.000 <sup>b</sup>	.064	.145	.060 (.090)	.032 (.048)
PSubMannerCI	.031	.068	.076	.013	.020	.018	.031	.016 <sup>b</sup>	.013	.077	.037 (.063)	.012 (.022)
PSubVoicingCI	.036	.047	.059	.023	.030	.024	.036	.008 <sup>b</sup>	.056	.093	.042 (.040)	.009 (.016)
<b>Phonological processes<sup>c</sup>: picture naming, word, and nonword repetition tasks combined</b>												
PTypSubCI	.045	.125	.099	.028	.069	.033	.053	.008 <sup>b</sup>	.074	.157	.071 (.075)	.028 (.044)
Fronting	.022	.244	.067	.000	.161	.000	.022	.000 <sup>b</sup>	.044	.178	.076 (.126)	.064 (.124)
Stopping of fricatives	.000	.030	.000	.000	.018	.000	.013	.000 <sup>b</sup>	.000	.053	.012 (.033)	.004 (.015)
Denasalization	.038	.077	.192	.038	.000	.038	.077	.000 <sup>b</sup>	.077	.038	.061 (.098)	.030 (.059)
Voicing	.029	.047	.029	.019	.010	.019	.067	.000 <sup>b</sup>	.019	.083	.033 (.054)	.007 (.016)
Devoicing	.045	.044	.088	.026	.046	.027	.008	.014 <sup>b</sup>	.089	.105	.051 (.048)	.011 (.022)
Gliding	.000	.000	.015	.000	.000	.015	.000	.000 <sup>b</sup>	.000	.000	.003 (.012)	.003 (.012)
PAtypSubCI	.053	.068	.076	.009	.063	.018	.040	.000 <sup>b</sup>	.036	.131	.051 (.080)	.034 (.049)
Backing	.011	.011	.023	.000	.013	.006	.023	.000 <sup>b</sup>	.029	.053	.018 (.036)	.010 (.024)
Abnormal stopping	.000	.000	.049	.025	.000	.000	.012	.000 <sup>b</sup>	.037	.000	.013 (.036)	.005 (.019)
H-zation	.022	.016	.016	.000	.000	.011	.005	.000 <sup>b</sup>	.000	.044	.012 (.029)	.003 (.008)
Nasalization	.000	.000	.000	.000	.005	.000	.005	.000 <sup>b</sup>	.000	.000	.001 (.004)	.001 (.005)
Dentalization	.057	.011	.057	.000	.046	.000	.011	.000 <sup>b</sup>	.000	.060	.025 (.058)	.015 (.028)
Lateralization	.005	.049	.005	.000	.036	.005	.005	.000 <sup>b</sup>	.000	.037	.015 (.031)	.017 (.034)
<b>Developmental levels of complexity<sup>d</sup>: picture naming task only</b>												
L1CI (/p t m n j/)	1.00	.86	.91	1.00	1.00	1.00	.90	1.00	.95	.90	.95 (.05)	1.00 (.01)
L2CI (/k/)	1.00	1.00	1.00	1.00	1.00	1.00	1.00	<b>.67</b>	1.00	1.00	.97 (.11)	1.00 (.00)
L3CI (/f s x h/)	1.00	.80	.80	.90	<b>.70</b>	1.00	.90	.90	.80	1.00	.88 (.10)	.99 (.03)
L4CI (/v (b d)/)	1.00	.92	1.00	.92	1.00	.92	1.00	<b>.75</b>	.92	.92	.93 (.08)	.99 (.04)
L5CI (/l r/)	.90	<b>.60</b>	.90	.90	<b>.40</b>	1.00	.80	.90	.80	.90	.81 (.18)	.93 (.13)
<b>Auditory discrimination<sup>e</sup></b>												
Words (% correct)	69	—	67	75	58	86	—	83	83	78	74.9 (9.6)	83.7 (15.0)
Nonwords (% correct)	53	—	89	92	50	97	—	92	75	78	78.3 (18.1)	78.9 (14.4)
<b>Oral motor skills</b>												
OMMA <sup>f</sup> , iso.	100	69	69	700	77	100	96	85	85	100	88.1 (12.9)	97.0 (4.1)
OMMA, seq.	83	67	72	70	83	100	78	78	72	100	80.3 (11.6)	93.5 (6.4)
OMMA, seq. fast	70	60	70	70	60	90	70	70	50	70	68.0 (10.3)	88.8 (12.5)
OMMA, overall	89	67	70	85	76	98	86	80	75	95	82.0 (10.4)	94.4 (5.6)
DDK <sup>g</sup> score	1	0	0	0	1	1	0	—	1	0	—	—
DDK judgment	2	1	2	2	3	2	2	—	2	2	—	—

Note. FAS2, FASD3, and FASD8 (underscored) are the cases with cases of confirmed mild hearing loss, whereas no information on hearing status is available for FASD1 and FASD10. All other cases feature no recorded hearing loss.

<sup>a</sup>Results are based on picture naming and word repetition only as this participant did not complete the nonword repetition task. <sup>b</sup>Phonological features: proportions of substitutions of syllable-initial consonants divided into substitutions of place of articulation, manner of articulation, and voicing (PSubPlaceCI, PSubMannerCI, and PSubVoicingCI; see also Table 3). <sup>c</sup>Phonological processes: proportions of substitutions of syllable-initial consonants divided into typical and atypical processes (substitution processes that are typical for a speech delay [PTypSubCI] and substitution processes that are indicative for a speech disorder [PAtypSubCI], respectively) as well as broken down into separate substitution processes (see also Table 3). <sup>d</sup>Developmental levels of complexity: proportions of correctly produced consonants according to the developmental levels of complexity for Dutch (Beers, 1995). The boldfaced numbers indicate that they fall below the 75%-correct criterion meaning that the corresponding speech sound category is considered not to be acquired yet. <sup>e</sup>Psycholinguistic Assessment of Language Processing in Aphasia (Bastiaanse et al., 1995). <sup>f</sup>Oral motor movement assessment (Erlings-van Deurse et al., 1993). <sup>g</sup>Diadochokinesis ([pataka]): Computer Articulation Instrument (Maassen et al., in press).